POST-APOPLECTIC TREMOR (SYMMETRICAL AREAS OF SOFTENING IN BOTH LENTICULAR NUCLEI AND EXTERNAL CAPSULES.)*

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The features of interest in the case to be reported are, clinically, a tremor of the right arm, resembling post-hemiplegic tremor, and ataxia of the arms and legs, and, pathologically, the presence of symmetrical lesions of the putamen.

The patient, a man of 58, was admitted to the Philadelphia Hospital on April 20, 1903, his chief complaint being difficulty in walking. The family history, as well as the previous history, were unimportant. Owing to the mental condition of the patient, a reliable account of the onset of his disease was not obtainable, his answers being frequently contradictory.

He stated when admitted that his present trouble had begun 3 years previously. He complained of pain in the head, weakness and numbness of the legs, and weakness of the right arm. At this time nothing of moment was noted excepting that the right knee jerk was increased, some ataxia was present in the upper limbs, and the patient had a tendency to lose his balance if he turned suddenly while walking. There was no Babinski reflex, and no apparent weakness in any of the limbs.

He came under the care of one of us (Dr. Potts) in April, 1906. At that time the peculiar movements of the right arm attracted attention. These consisted of alternate flexion and extension at the elbow and wrist, the hand being supinated. The movements were almost constant, only ceasing for a time if his attention was distracted. At this examination he told a somewhat different story than at first, saying that at the beginning of his disease he noticed weakness of the legs, the left being weaker than the right, and that these symptoms dated back eleven years, following what he called "a cold." Six years later the right arm became affected, the disorder consisting at first of a difficulty in writing. Examination at this time showed a somewhat ataxic gait, which was more apparent with the shoes off. The right leg was held more stiffly than the left. Station, with the eyes closed, was poor, and there was also marked ataxia of the arms, especially the right. There was apparently no weakness of the arms. The legs appeared to be somewhat weaker than normal. The biceps, triceps, and wrist jerks were not marked. The knee-jerks were prompt, the left being more so. The plantar reflex gave a normal response. Examination of the eyes showed a sluggish response to light, but otherwise they were normal.

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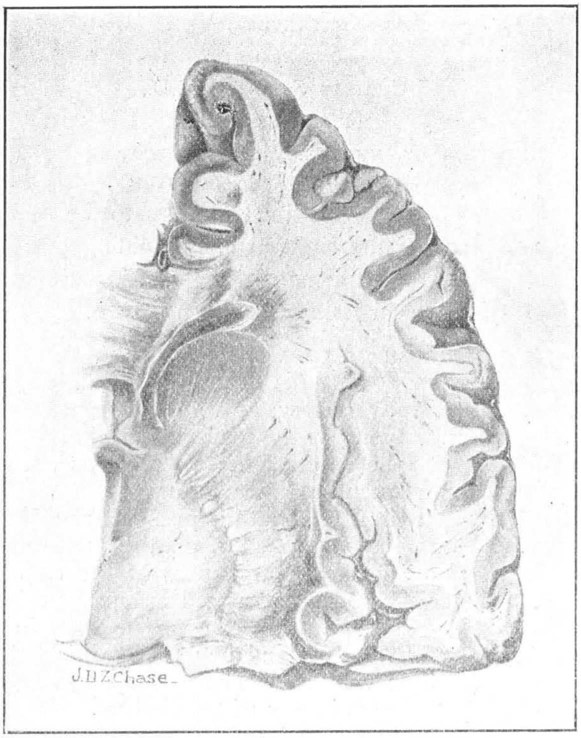

Fig. 1. Linear area of softening in external capsule on right side. Upper limit. (Macroscopical section.)

The brain and spinal cord were turned over to Dr. William G. Spiller, who gave them to one of us (Dr. Rhein) for examination. The specimens were hardened in formalin.

Horizontal sections of the brain on the right side revealed the presence of an area of softening which first made its appearance a short distance above the lenticular nucleus, and was situated in the external capsule, *i. e.,* between the claustrum and the internal capsule. This area of softening was linear in shape, and measured at this level 2.5 cm. in length, and about 1 to 2 mm. in width. While it lay directly external to the fibers of the internal capsule, it did not cause any degeneration of these fibers.

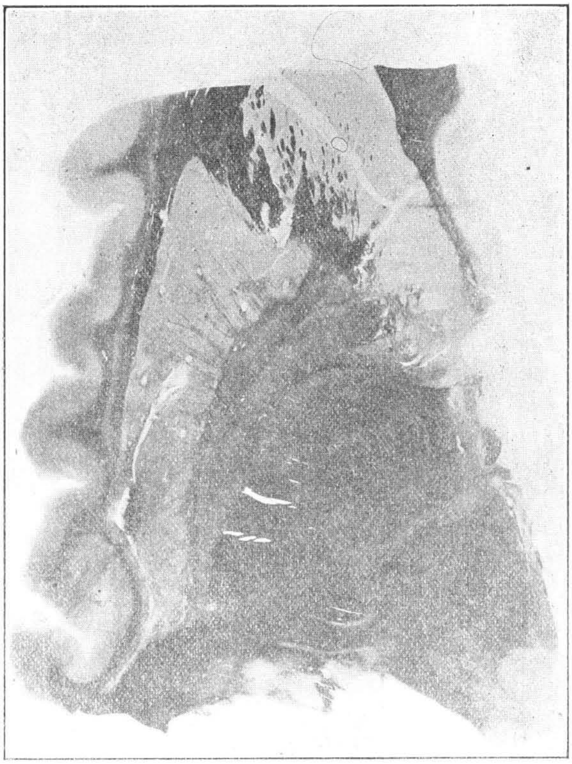

Fig. 2. Area of softening extending into the middle portion of the right putamen (microscopical section, section reversed in photograph).

The focus extended downward in a vertical direction to a position corresponding to the lower limit of the island of Reil. At the lower level of the lenticular nucleus it destroyed in part the external portion of the putamen in its posterior third. At the level at which the fasciculus of Türck is seen ("temporale Brückenbahn"—Obersteiner) these fibers were in part destroyed. The anterior commissure was intact, as were also the fibers of the internal capsule. The fibers of the external part of the foot of the peduncle, corresponding to the situation of the fasciculus of Türck, stained poorly. The red nucleus appeared to be intact.

On the left side of the brain there was a similar lesion which began at a somewhat lower level than the one on the right side, its upper limit being at about that level where the lenticular nucleus is first seen in horizontal sections. It was situated in the same relative position as that of the cavity on the right side, occupying the region of the external capsule between the claustrum and the putamen, and measured 3.5 cm. in length, and 10 mm. in its widest portion. It extended downward in a vertical direction, and disappeared a little above the level of the lower border of the island of Reil. The posterior third of the putamen in its lower part was in large measure occupied by this focus of softening. The fibers in the retro-lenticular region were more or less involved.

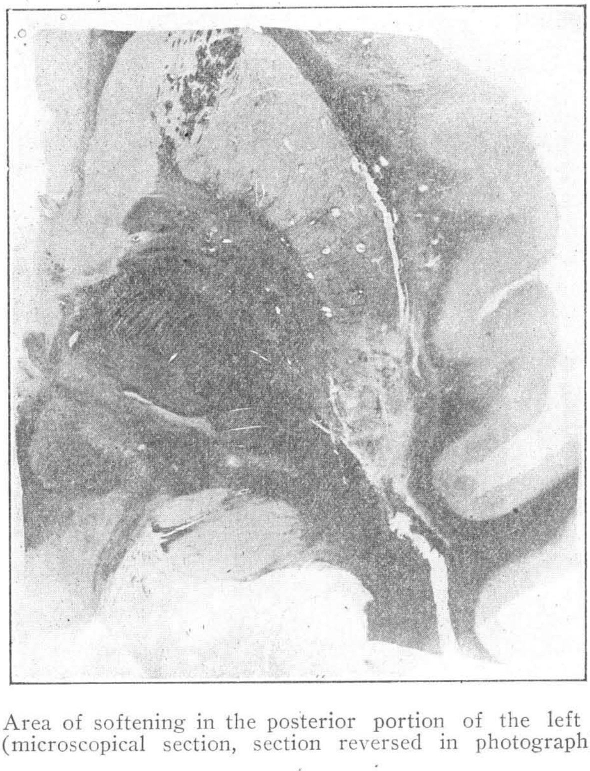

Fig. 3. Area of softening in the posterior portion of the left putamen (microscopical section, section reversed in photograph).

The fibers in the internal capsule were not degenerated. The red nucleus appeared to be normal.

Certain fibers passing inwardly and apparently through the lenticular nucleus toward the thalamus, stained well on both sides.

In sections from the pons, medulla oblongata and spinal cord the nerve fibers stained normally. There was no evidence of descending degeneration in the pyramidal tracts.

The cells of the anterior horns in the cervical and lumbar regions, stained by hemalum and acid fuchsin, showed no marked change.

Sections from the cerebellum showed an unusual vascularity of the dentate nucleus on both sides. The vessels were numerous and somewhat thickened, and surrounded by a slight round-cell infiltration, and there was also some fresh hemorrhage about some of the vessels.

To summarize the pathological findings in this case, there was an area of softening in each putamen, which caused degeneration in the fasciculus of Türck, on the right side. There was no degeneration in the motor fibers of the internal capsule, the pons, medulla oblongata or spinal cord.

Associated with these pathological findings, the tremor of the right arm and the ataxia of the arms and legs are worthy of some discussion.

In regard to the first of these symptoms, namely, the involuntary movements of the right arm, which resembled a post-hemiplegic tremor, it is interesting to note that lesions of the lenticular nucleus have been thought by some to cause post-hemiplegic chorea. Among the first to describe cases of this nature was Demange,[1] who, in 1883, reported 11 cases with autopsy, in 6 of which the lenticular nucleus was the seat of disease.

Sander,[2] in 14 cases of athetosis collected from the literature, found the corpus striatum involved in 7 cases. In the case described by Grasser and Rauzier[3] the lenticular and caudate nuclei, and the internal capsule were partly destroyed by an area of softening.

The putamen was symmetrically diseased in one case described by Hebold,[4] and in a second case the claustrum was also involved on both sides. He quotes Nothnagel who collected cases of athetosis and hemichorea, in which the lenticular nucleus and the internal capsule were involved. In Anton's[5] case the choreiform and athetoid movements were on the same side as the lesion in the putamen, and in Jacob's[6] case the contralateral lenticular nucleus was the seat of the disease.

Many cases have been cited in which, besides the lenticular nucleus, the thalamus has also been involved. This was true in 50 out of 78 cases collected by Bidon.[7] In about 100 cases studied by v. Monakow,[8] in 70 per cent. to 80 per cent. there was also lesion of the internal capsule.

On the other hand, bilateral lesions of the lenticular nucleus may occur without causing tremor, as in Edinger's[9] case, and in 2 cases of Hebold.

If foci of softening or other lesions of the lenticular nucleus cause tremor, it is hard to reconcile entirely the findings in our case with this statement, as tremor was only present in the right arm, while the putamen was diseased on both sides. Of interest in this connection is the opinion of Hebold who believed that a different explanation for the motor disturbances than injury to the putamen, must be looked for, at least as far as the outer portions are concerned.

It should be remembered, however, that in our case the lesion caused much greater destruction in the left putamen than in the right, involving, in the former, a large part of the posterior third of the putamen, while on the right side the lesion was linear in shape and occupied the external portion, the destruction of the putamen being less extensive. It may be that the lesion on the right side was not sufficiently large to cause disturbances of motion, while the damage to the left putamen, being much more extensive, was sufficient to give rise to a tremor in the right arm. This is of course somewhat theoretical.

At least the findings in our case permit the statement that bilateral lesions of the putamen do not always cause bilateral disturbances of motion.

The pathology of post-hemiplegic disturbance of motion has not been definitely established, and the views held upon the question are far from uniform. On the whole, however, the weight of evidence is in favor of placing the lesion in one of several localities, and preferably in the thalamus, or in the pons, in the neighborhood of the superior cerebellar peduncles and red nucleus. Many observers have, however, described tremors, choreiform movements and athetosis post-hemiplegic in type, in organic disease of the cerebellum, medulla oblongata, cortex and spinal cord.

Lesions of the thalamus have been regarded by many observers as the chief cause of these movements. Since Charcot,[10] in 1873, reported 3 cases with autopsies, in which the lesion involved the posterior portion of the optic thalamus, caudate nucleus, and corona radiata, a number of cases of this character have been described, namely, those of Stephan,[11] Sander, Gowers,[12] Bidon, Kahler and Pick,[13] Raymond,[14] Bischoff,[15] and Leyden,[16] who found various lesions affecting the thalamus, with or without involvement of the surrounding tissues.

In 100 cases studied by v. Monakow, it is stated that, in the majority of cases the lesion was found principally in the thalamus. In another group of cases the retrolenticular portion of the internal capsule and the thalamus were the seat of disease, while in the remainder of the cases the posterior portion of the corona radiata and the lenticular nucleus were involved.

Raymond, however, on the contrary, noted in 35 cases of hemiplegia, involvement of the thalamus in only 4 cases, and Sander believed that there were only a few cases in which the thalamus alone was involved. Moreover, the thalamus and the internal capsule were affected together in 20 cases in the literature studied by Stephan.

The relation of the post-hemiplegic movements to the cerebellar peduncles, the tegmentum, and the red nucleus, was probably first described by Benedickt[17] who, in 1874, reported 22 cases of hemiparesis and chorea caused by a lesion in the crus cerebri, at the height of the oculomotor nucleus. Halban and Infeld[18] added to these 2 cases of their own, in one of which a tuberculous focus had destroyed the red nucleus in large part, extending almost to the decussation of the superior cerebellar peduncles. Henoch[19] also observed a case with tremor, hemiplegia, and crossed oculomotor palsy, in which there was found a tuberculous lesion of the pons and corpora quadrigemina, and Bergen[20] collected 10 cases in which the cerebellum and superior cerebellar peduncles were destroyed, causing incoördination identical with hemichorea on the same side as the lesion.

The superior cerebellar peduncles, the red nucleus and the tegmentum were diseased in a case reported by Bonhoffer,[21] who believed that choreiform movements and athetosis were due to a lesion of the centripetal fibers extending from the cerebellum to the cerebral cortex, giving rise to a functional disturbance of the motor cells of the cortex. He advanced three reasons for accepting this view, namely, (1) the constant presence of a lesion of the superior cerebellar peduncles, or its projections into the subcortical ganglia; (2) the hypotonia of the musculature; (3) the disturbance of voluntary motion in chorea. He thought it probable that, in consequence of a more or less complete interruption of the fibers, the centripetal impulses only in part reached the cortex, while others passed along the centrifugal fibers and gave rise to automatic movements the character of which depended upon the nature and extent of the lesion.

Pineles[22] reported 2 cases of athetosis which he considered of cerebellar origin, and cited the case of Oliver, in which there was a glioma of the vermiform process in a case presenting tremors of the arms and legs. He cited also Menzel's case of choreiform movements with atrophy of the cerebellum, a similar case of Meynert's, Ceni's case of atrophy of the right hemisphere of the cerebellum, and right cerebellar peduncle, with a hemorrhagic focus in the left red nucleus, and Sander's[4] case in which there were choreiform movements on the right side, caused by a gliosarcoma in the right cerebellum, destroying the corpus dentatum. Pineles concluded that in many cases choreiform movements and athetosis were caused by disease of the cerebellum, or the cerebellar peduncles. Other cases of a similar nature have been reported by Kirschoff,[23] Bonhoffer, Huppert,[24] Muratoff,[25] and Hammarberg.[26] In Hammarberg's case the movements were pendulum-like.

Lesions in the medulla oblongata, causing hemichorea, were described in Bidon's cases (collected from the literature), in v. Monakow's case and in the cases of Fropier,[27] and Broadbent (in v. Monakow) and Henoch.

Cortical lesions have been described as causing athetoid and allied movements in the 2 cases of Demange, and those of Balfour,[28] Beach,[29] Korella,[30] and Hudovernig.[31] Some of these cases are, however, so meagerly reported as to be of little value.

Opposed to this is the statement of Exner (in Frey) who collected 167 cases of cortical lesion without one instance of post-hemiplegic tremor. In this connection, as bearing upon the irritation of the motor tracts as causing post-hemiplegic movements, must be mentioned the cases in which the internal capsule has been the seat of the lesion, as illustrated by the case of Demange, and those collected by v. Monakow, Bidon (29 cases), and others.

Finally, Eisenlohr[32] and Anton describe cases in which spinal lesions were mentioned as causing the movements under discussion.

Several theories have been advanced to explain the occurrence of these movements, among the first of which was that of Charcot, who believed that they were due to disturbances of a "chorea-bundle," a view long since abandoned.

Kahler and Pick, who concluded that cysts or areas of softening in the thalamus, with or without involvement of the internal capsule, give rise to hemichorea and other disturbances of motion, believed that these were due to irritation of the pyramidal fibers, a view also held by Sander,[39] Demange, Kolisch,[33] Hudovernig, and Nothnagel.[34] Frey[40] and others, however, object to this theory on the ground that the usual result of irritation of the pyramidal tracts is to cause convulsions and spasms; that hemiplegia is a frequent occurrence, while choreiform movements are rare; that lesions removed from the motor tracts cause these symptoms; that paresis is absent in some cases; that it is improbable that complicated movements should remain circumscribed if thus explained (v. Monakow); that the head is affected in deep-seated lesions, and that these symptoms are infrequent in lesions of the lenticular nucleus (Frey). Furthermore, it is advanced by some authorities that cortical lesions rarely cause post-hemiplegic movements, although the cases of Demange controvert this statement.

Von Monakow does not subscribe to the view held by Kahler and Pick and others, that these movements are the result of irritation of the pyramidal fibers which is conveyed to the cells of the anterior horns of the cord, but in his opinion the tissues surrounding the lesion, as in the subthalamic area, the tegmentum and the pons, become irritated, and that this irritation is transmitted to the motor cortex.

Bonhoffer, in explaining the symptoms, believed that the different choreiform movements are caused by alterations of the impulses which normally pass to the cerebral cortex through the tegmentum. As already stated, he believed that it was plausible that impulses pass centripetally to the cerebral cortex in part only while others pass directly in the centrifugal motor fibers, and give rise to automatic movements.

According to Frey and Stephan, the thalamus is a coördinating center which, when disturbed, gives rise to post-hemiplegic disturbances of motion. Frey believed that the cause of these movements is to be found in lesions of the thalamus and hypothalamic region.

Anton held that automatic associated movements originated in the posterior part of the optic thalamus and its connections, while the corpus striatum inhibited these movements. Therefore, lesions of these ganglia cause a lowered inhibition, and in consequence an increase of the automatic movements, or chorea.

Halban and Infeld believed that fibers going through the red nucleus have to do with a complicated mechanism, and that disturbance of these fibers gives rise to the automatic movements.

According to Sander, disease of the thalamus was the cause of athetosis in his case, and he expressed the opinion that lesion of the motor tracts causes in part of the fibers incomplete interference with the passing of nerve impulses, and that the accumulation of these impulses centrally from the lesion, *i. e.*, in the cortical cells, becomes so intense from time to time as to overcome the resistance at the point of the lesion, giving rise to movements of a rythmical nature.

Pineles inclined to the belief that while these movements may be associated with irritation of the pyramidal fibers, many of the facts speak for the relation of chorea to the superior cerebellar peduncles. He believed that the choreiform movements were the result of loss of function (an "ausfall" symptom).

One is struck, in studying the cases and the various theories above cited, with the fact that the majority of lesions described affected centers or fibers which are connected directly or indirectly with the cerebellum, and it does not seem improbable that, in some way not perfectly clear, the function of the cerebellum, at least as far as co-ordination is concerned, may be disturbed, and, as a result, give rise to choreiform movements, athetosis and tremor, which, according to some observers at least, may be looked upon as allied disturbances of motion.

The findings in our case may add weight to this view, if we are correct in assuming that the lenticular nucleus is associated with the cerebellum by fibers which pass from the lenticular nucleus on one side, to the contralateral cerebellar hemisphere by way of the ansa lenticularis.

Obersteiner[35] believes that the lenticular nucleus is connected with the lower olive on the same side, and the opposite cerebellar hemisphere in this way, and Mills[36] states that the corpus striatum is connected with the cerebellum through the red nucleus, the pontine nuclei, and olives, and the other basal ganglionic deposits.

According to Dejerine[37] part of the fibers of the ansa lenticularis penetrate into the antero-internal part of the capsule of the red nucleus, which is composed of fibers from the superior cerebellar peduncles.

These facts justify the assumption, to some extent, that the lenticular nucleus may have some function relating to co-ordination through its connections with the cerebellum which it may have at least indirectly, if not directly, by reason of its relation with the red nucleus. If, then, as a result of a lesion of the lenticular nucleus, the fibers associated directly or indirectly (through the red nucleus or otherwise) with the cerebellum be interrupted, and thus the functions of the cerebellum be impaired, may not disturbances of motion arise expressing themselves in involuntary movements or ataxia?

This brings us to the consideration of the ataxia of the arms and legs which was present in our case, and offers a possible explanation for this symptom, which is not explained in any other way by the pathological findings already described.

The tremor in our case, we believe, was not hysterical, as the patient exhibited no other stigmata of hysteria, although of course tremor may be the only manifestation of hysteria, as in the case described by Mitchell and Spiller.[38]

IN CONCLUSION, we believe:

1. That the lesion in the left lenticular nucleus may have been responsible for the tremor of the right arm in our case, and the association of unilateral tremor with bilateral lesion may be explained by the fact that the lesion in the right putamen was not extensive enough to set up sufficient irritation to cause this symptom on the left side.

2. That post-apoplectic disturbances of motion may be due to lesions of the lenticular nucleus, the optic thalamus, and the pons in the region of the superior cerebellar peduncles, and of the cerebellum, the cause being, in all instances, a disturbance of co-ordination. When the lesion is extra-cerebellar the cause of this disturbance is possibly an indirect result of destruction of fibers related to the cerebellum directly or indirectly through the red nucleus.

3. That the ataxia in our case may have been indirectly of cerebellar origin.

It must not be forgotten, in drawing conclusions from an analysis of the above cited cases, that there are on record instances in which lesions of the posterior portion of the thalamus (**"hintere Sehhügelgegend"**), **red** nucleus, superior cerebellar peduncles, dentate nucleus, and cerebellum have occurred without causing athetoid and choreiform movements (v. Monakow).

We gladly express our thanks to Dr. W. G .Spiller for valuable assistance in the study of the specimens.

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